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Florid cystic endosalpingiosis of the uterine subserosa: A case report

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Dear Editor:

Florid cystic endosalpingiosis (FCE) is a rare type of endosalpingiosis that presents as a mass-like lesion. It may rarely involve the uterus and present as a cystic or tumor-like mass. Herein we report an unusual case of FCE. The patient is a 43-year-old woman presented with pelvic cystic mass, which was diagnosed as an ovarian cyst by ultrasonic examination, and diagnosed as a uterine leiomyoma with cystic degeneration by laparoscopy. Pathologic findings show FCE of the uterine subserosa. Preoperative diagnosis of uterine cystic endosalpingiosis is usually difficult. Thus, awareness about this condition in clinicians may help in preventing misdiagnosis and overtreatment.

A 43-year-old woman, gravida 6 para 1, presented a cystic formation at the left adnexal area. The formation was first detected 4 years earlier by transvaginal sonography (sized 3.0cm × 4.2cm) during routine gynecologic check-up. Considering that the lesion is benign and absence of additional clinical symptoms, the patient selects for follow-up observation. During the latest check-up, a slight increase in size of the cystic formation (3.7cm × 5.0cm) was noticed and the patient reported to have suffered from chronic pelvic pain for 2 years without vaginal bleeding or menstrual disorder. The patient had no significant past medical or surgical history, nor any family history. Transvaginal sonography showed a single, roughly circular, unilocular mass with homogeneous cystic content, measuring 3.7cm × 5.1cm, located in the left adnexal area. A pelvic examination revealed a large tender mass fixed in the left uterine corner. Serum CA199 was slightly increased to 39.3U/mL (normal range 0–37U/mL), while CA125 and biochemical parameters were within the normal range.

Laparoscopy revealed a large cystic and solid mass protruding from the left uterine corner. The mass was located beneath the uterine serosa and slightly invaginating the myometrium, which was similar to a subserosal leomyoma (Fig. 1A). Both ovaries and fallopian tubes appeared normal. No endometriotic lesions were found in pelvis. The mass was resected via laparoscopic myomectomy. The clinician was perplexed as to the origin and nature of the cyst. Intraoperative diagnosis was uterine leiomyoma with cystic degeneration.

After uneventful laparoscopic resection, the patient quickly recovered and expressed relief of her pelvic pain. The patient did not receive any adjuvant treatment and no remarkable symptoms were noted through her follow-up until now.

In pathological examination, the specimen was a gray to white colored, solid and cystic mass, having a smooth outside surface, measured about 5 cm in the largest diameter. The unilocular cyst was filled with clear serous fluid in transverse section, the cyst wall was 0.7 cm–1.0 cm thick (Fig. 1B).

Microscopically, the cyst was lined with a singular or stratified layer of ciliated tubal-type epithelium. These cells showed bland nuclear features, without mitotic figures. The cyst wall was composed of loose connective tissue surrounded by normal myometrium. There were no endometrial stromal-like or previous hemorrhage areas near the epithelial lining and no endocervical-type mucinous epithelium. Mixed cell types of columnar and ciliated cells with intercalated (peg) cells were noted at higher magnification. The intercalated cells possessed a clear cytoplasm (Fig. 1C). A few tiny glands lining tubal epithelium were found beneath the epithelium. The immunohistochemical staining showed that the lined epithelial cells were positive for WT-1 (Fig. 1D), EMA, CK7, estrogen receptor (ER), and progesterone receptor (PR). The stroma beneath the epithelium was loose connective tissue, positive for vimentin, the tubal epithelium also stained strongly for vimentin. No CD10 positive cells were found. The final pathologic diagnosis is FCE of the uterine subserosa.

Endosalpingiosis is one of the müllerianosis triad. According to WHO definition, "Endosalpingiosis" is the presence of glands lined by benign tubal-type epithelium outside of the fallopian tube.[1] It was first described by Sampson in the year 1930.[2] Most cases of endosalpingiosis are asymptomatic, while it may sometimes present with nonspecific symptoms including pelvic pain, infertility, abnormal uterine bleeding, and/or urinary symptoms, making preoperative diagnosis nearly impossible. The finding is typically incidental in women undergoing surgery for pelvic pain, infertility, urinary symptoms, or a pelvic mass, and the biggest obstacle in its diagnosis is that it can only be confirmed by histopathological examination.

Endosalpingiosis is a more important disease without given enough attention, which has two extremes. On one hand, it presents as a few glands lined by tubal-type epithelium under microscopy, while on the other hand, it undergoes massive cystic change and presents tumor-like manifestation. This tumor form of endosalpingiosis is defined as florid cystic endosalpingiosis (FCE) by Clement and Young in 1999, sixty-nine

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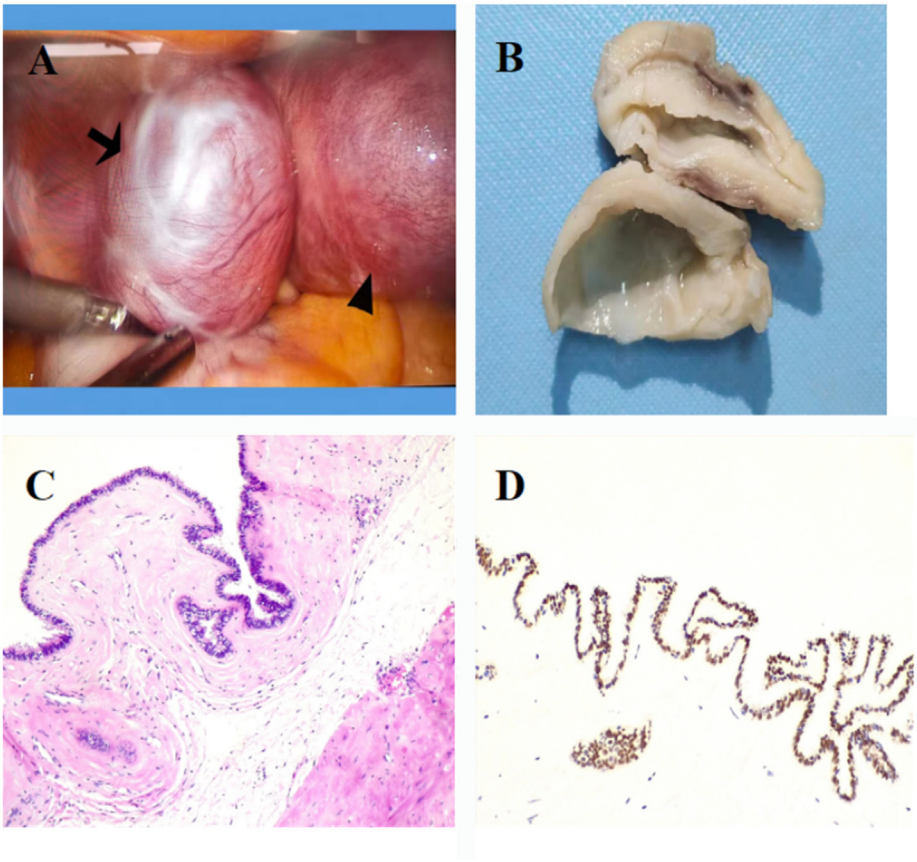

Fig. 1. A: Laparoscopy revealed a large cystic and solid mass (arrowhead) protruding from the left uterine corner (arrow), beneath the uterine serosa and slightly invaginating the myometrium. B: The specimen, gray to white colored, measured 5cm × 5cm × 5cm; cyst wall thick 0.7cm to 1.0 cm. C: Microscopic slide, loose connective tissues surrounded by normal myometrium and lined with a layer of ciliated tubal epithelium. The intercalated cells possessed a clear cytoplasm. D: Immunohistochemistry, the lined epithelial cells were positive for WT-1.

years after the first report of the common type of endosalpingiosis.[3] Clement and Young reported four cases of FCE as a series, highlighting its characteristic of mimicking a neoplasm. The morphological features of our case most closely resemble those described by Clement and Young and designated as endosalpingiosis with tumor-like manifestation.[3]

Endosalpingiosis has been reported in various organs, within and outside of the reproductive system. Endosalpingiosis was most commonly found on the ovary (36.8%), followed by the fallopian tube (21.3%), omentum (8.8%), and uterus (6.6%); other sites included uterovesical pouch, peritoneum, lymph nodes (pelvic, parametrial, obturator, external iliac), cervix, bladder, appendix, abdominal wall, pouch of Douglas, small bowel and sigmoid colon.[4] Cases of endosalpingiosis outside of the reproductive system are excessively rare. The clinical presentation of endosalpingiosis vary widely depending on the involved organs.

FCE rarely appears in the uterus. Recently, Peixinho et al. reviewed all the published cases of uterine FCE and found only thirty-two cases described in the literature.[5] FCE of the uterus can be categorized into two types based on their location: intramural or sub-serosal.[6,7]

Cystic tumors arising from the uterus are uncommon. Of those tumors, leiomyoma with cystic degeneration is the most common diagnosis after surgery. Another common cystic tumor arising from the uterus is cystic adenomyoma, which locates within the myometrium. This rare entity has been described as a focal form of adenomyosis.[8]

When differentiating FCE with other uterine lesions, histological examinations and special immunostaining is crucial. Leiomyoma with cystic degeneration is a pseudocyst, not epithelia lining; cystic adenomyoma lines with the endometrial epithelium and has endometrial stroma associated with hemorrhagic content. The diagnosis of endosalpingiosis depends on pathological findings of the existence of tubal-like epithelium containing three types of cells: ciliated columnar cells, non-ciliated columnar secretory mucous cells, and so-called intercalary or peg cells in an ectopic location. WT-1, the Wilms' tumor gene, plays a big part in diagnosing endosalpingiosis. In normal female genital tissues, ovarian surface epithelium and tubal epithelium express WT-1, while cervical or endometrial epithelia do not.[9,10] Further, immunohistochemical analysis can clearly distinguish cystic endosalpingiosis from cystic adenomyoma, in which its endometrial stroma shows strong positive for CD10.

We have a limited understanding of the etiology and clinical significance of endosalpingiosis. Recently, Sunde et al.[11] and Essenlen et al.[12] have demonstrated that endosalpingiosis is much more common than previously recognized. Thus, the clinical significance of endisalpingiosis should be a continued area of research. Several hypotheses were developed, including celomic metaplasia, implantation during surgery, and müllerianosis. The most accepted theory of pathogenesis is metaplastic change of coelomic cells into tubal-like epithelium.[13] The metaplastic theory may explain the reason why some cases have been reported in the absence of gynecological diseases or surgical history, and why lesions may have multiple müllerian components reflecting lesion differentiation.

Therefore, we should pay more attention to this rare type of benign lesion. Awareness of endosalpingiosis is necessary for diagnosis and treatment of this condition. It is necessary to keep this benign condition in mind both in clinical practice and surgical pathology. The fact that endosalpingiosis could be misdiagnosed as malignancy[13,14] and some cases of endosalpingiosis had a concurrent gynecological malignancy give more complexity to the diagnosis.[13] Although it is a clinically unexpected finding, it should be considered in the differential diagnosis of a uterine cystic mass. Fully understanding the histological features and give awareness of this condition will facilitate accurate diagnosis and to prevent overtreatment. Most patients with endosalpingiosis underwent a hysterectomy with salpingo-oophorectomy, but if possible, conservative treatment should be considered.[15,16] We here highlight the application of

laparoscopy, which could resect the cyst and make diagnosis with minimal invasion.

In conclusion, FCE is a rare-type endosalpingiosis seldom with uterus involvement. We have encountered this case for the first time in our practice. Proper histopathological diagnosis of the condition is crucial. Clinician and pathologist should understand this disease in order to diagnose correctly and avoid overtreatment.

Ethical approval and consent

The patient hospitalized and performed the removal of the cyst in our hospital (Beijing Wuzhou Women and Children's Hospital) and was approved by the Institutional Review Board of our hospital. The patient was fully recovered and successfully discharged. The patient was informed with the rarity and worth of reporting the case, and we promised there would be no personal information present in the report. The patient consented to publish the case.

Declaration of competing interest

The authors declare no conflict of interest.

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